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CASE REPORT
Hand/Peripheral Nerve

A Toe Keloid after Syndactyly Release Treated with Surgical Excision and Intralesional Steroid Injection

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Summary: A keloid is a benign fibroproliferative disease of unknown etiology. Although it is common among Asians, the development of keloid on the foot is rare. We experienced a case of a keloid which arose on the foot of a 4-year-old boy after the surgical release of syndactyly. He had congenital cutaneous syndactyly of the third and fourth toes. After the reconstructive operation was performed when the patient was 2 years old, the wound became hypertrophic and grew to 37×37×8 mm. After the diagnosis of keloid based on a pathological examination, the keloid was resected completely. The web was reconstructed with a planter rectangular flap, and the skin defects were covered with a full-thickness skin graft. After the operation, we administered 5 intralesional steroid injections. Finally, the keloid was diminished 2 years after the operation. *(Plast Reconstr Surg Glob Open 2014;2:e186; doi: 10.1097/GOX.0000000000000152; Published online 24 July 2014.)*

A keloid is a fibrous proliferative benign tumor that invades the adjacent normal skin beyond the boundaries of the initial scar and is often resistant to treatment. Patients complain of a disfiguring scar, itching, and pain of the lesion. The most common sites of keloids are the anterior chest, shoulder, earlobe, and upper arm. However keloid formation on the fingers and toes is very rare.

We herein present a case of toe keloid that developed after the web release operation for syndactyly, which was cured completely by surgical excision and a skin graft, combined with intralesional steroid injections.

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CASE REPORT

A 4-year-old Japanese boy had been born with cutaneous complete syndactyly of the third web of the left foot. He underwent a web release operation at the age of 2. The web was reconstructed using the dorsally based flap, and the residual skin defects were covered with a split-thickness skin graft from the medial malleolus. The postoperative course was uneventful. Two months later, the scar became red and hard with itching. Compression treatment with a sponge was started, and adhesive tape containing a steroid was applied. Despite these additional treatments, the scar became hypertrophic and enlarged.

When the patient came to our hospital, the third and fourth toes were fused completely, and a red and hard mass of 37×37×8 mm appeared on the dorsal side of the fused toes (Fig. 1). He complained of having problems putting on his shoes.

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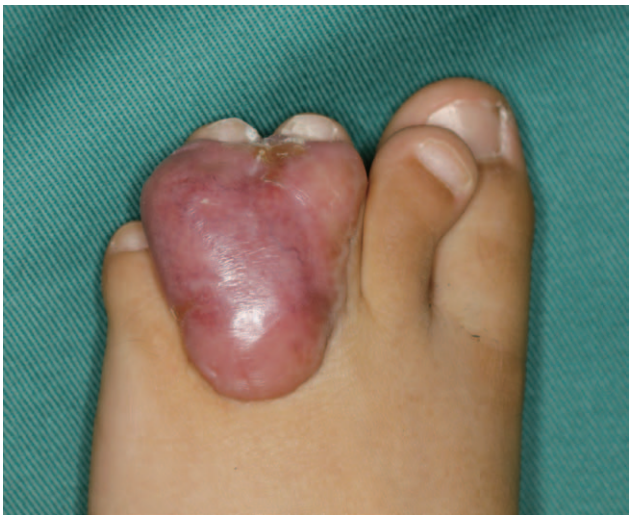

Fig. 1. A keloid on the third and fourth toes. The third and fourth toes were fused completely.

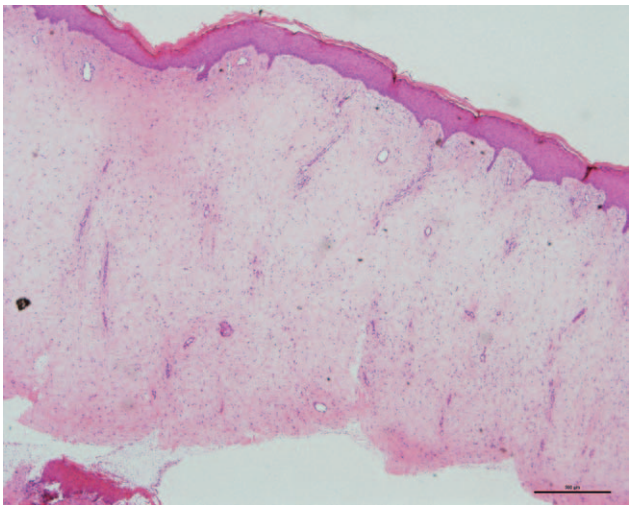

Fig. 2. Hematoxylin and eosin staining. A biopsy specimen was diagnosed as a keloid. Bar: 500 μm.

The scar at the donor site was mature and flat, and no other keloids were detected. The preoperative radiographic examination did not reveal any macrodactyly of the affected side of the foot. A biopsy was first performed to obtain a definitive diagnosis. A biopsy specimen demonstrated that the epidermis was flattened, and the dermis was homogenous and edematous from the papillary layer to the reticular layer. Dilation of lymph ducts and sparse infiltrations of inflammatory cells were detected (Fig. 2). No intracytoplasmic inclusions were detected. The immunohistochemical analyses revealed that cytokeratin, desmin, CD34, D2-40, and β-catenin were not expressed, whereas the staining for collagen type 1 and vimentin was positive. Finally, the specimen was diagnosed as a keloid.

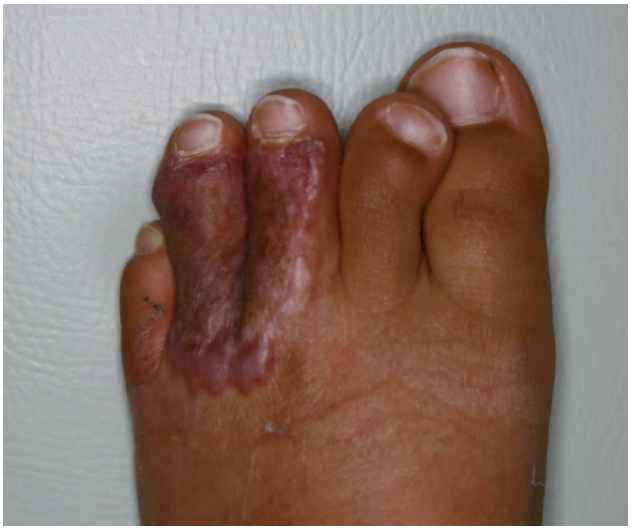

Fig. 3. Three months after the operation. The keloid recurred on the edge of the skin graft.

Surgical Procedure

We resected the keloid completely, the third web was reconstructed with a plantar-based rectangular flap, and the lateral toe defects were covered with full-thickness skin grafts from the abdomen. The postoperative course was uneventful. The skin grafts were all successful, and the wound healed completely by postoperative day 11. On the 20th day after the operation, compression treatment with a sponge (Reston, 3M Co. Ltd., Tokyo, Japan) was started.

Postoperative Course

The margin of the skin graft became red and hard 2 months later. Although a steroid patch was applied at that time, the keloid recurred (Fig. 3). We then injected triamcinolone acetonide into the recurrent keloid 5 times (4mg per injection) during the 16 months after the operation. The keloid was cured completely by 2 years after the operation. After that, no recurrence has been detected for 1 year, without additional treatment (Fig. 4).

DISCUSSION

We experienced the large keloid of the toe that was cured completely with surgical treatment, followed by triamcinolone acetonide injection. Keloid formation on the toes and fingers is rare, and so far only 21 cases in 8 reports have been reported.[1–8] Therefore, we first suspected that the patient had a fibrous tumor, such as infantile digital fibromatosis. Fibromatosis is a benign fibroblastic tumor, and there are 2 main types that occur in infancy and childhood. The first type is similar to the adult lesions, which consist of nodular fasciitis, palmar fibromatosis, and abdominal fibromatosis, and the

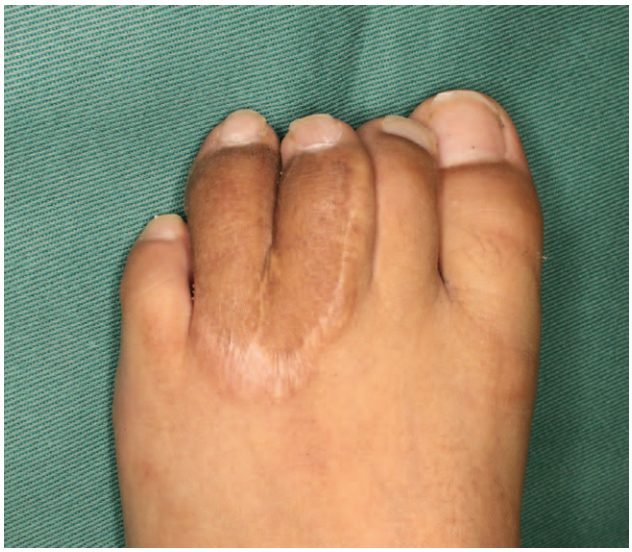

Fig. 4. Three years after the operation. No recurrence has been detected.

second type is specific to infancy and childhood, and we suspected that our case might represent one of the latter cases. However, the mass was finally diagnosed as a keloid based on the pathological findings.

Keloid formation after the reconstruction of syndactyly might be associated with macrodactyly and Proteus syndrome. Muzaffar et al[5] reported that 8 patients developed keloids out of total of 1008 syndactyly operations on 681 patients, and 7 of these 8 patients had the macrodactyly. Moreover, 2 of the 7 patients had Proteus syndrome, which is characterized by partial gigantism of the hands and/or feet, with overgrowth of fatty tissue, pigmented nevi, hemihypertrophy, subcutaneous tumors, skull anomalies, accelerated growth, and visceral anomalies. Our case did not show either macrodactyly or partial gigantism.

Various treatments for keloids have been described. We have previously treated keloids using surgical excision and postoperative irradiation as a standard treatment[9]; however, the radiosensitivity of the children is so high that hypoplasia following foot irradiation is inevitable. Therefore, intralesional steroid injections were substituted for radiation treatment as an additional postoperative treatment in the present case. Finally, 5 injections of triamcinolone acetonide over a period of 16 months brought about a complete cure. Onwukwe[9] reported the successful outcomes of using methotrexate in the treatment of keloids. Low-dose methotrexate is widely used for rheumatoid arthritis and is used in both adults and children. However, as the keloid in our patient was a local disease, we elected to use a local application to minimize the systemic effects.

CONCLUSIONS

We experienced a case of toe keloid that developed after the web release operation for syndactyly, which was cured completely by surgical excision and a skin graft, combined with intralesional steroid injections.

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